

Developmental absence of maxillary lateral incisors in the Portuguese population

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SUMMARY The aim of this study was to evaluate the prevalence and clinical manifestation of developmental absence of maxillary permanent lateral incisors in the Portuguese population.

The study group comprised 16 771 patients observed between 1993 and 2000 at the Faculty clinic. Two hundred and nineteen patients were found to have missing upper lateral incisors (131 females and 88 males, with ages ranging between 3 and 71 years), a prevalence of 1.3 per cent in this population. Absence of these teeth was bilateral in 44.7 per cent of the patients; of the unilateral cases, 33 per cent occurred on the right side and 21.9 per cent on the left side.

In children under 8 years of age ($n = 12$), two (0.26 per cent) also had developmental absence of the primary lateral incisors, one unilateral and the other bilateral.

Among the 121 subjects with unilateral developmental absence, the contralateral maxillary lateral incisor was found to be microdont in 57.1 per cent (right lateral 23 per cent, left lateral 34.1 per cent), suggesting the possibility that microdontia represents a variable expression of the same developmental defect that results in absence.

Introduction

Developmental lack of a tooth results from disturbances during the early stages of tooth development. A tooth is defined as developmentally missing if it has not erupted in the oral cavity and is not visible radiographically.

Hypodontia is a relatively frequent anomaly of the permanent dentition, with a reported incidence of between 3.5 and 8.8 per cent, when third molars are excluded (Horowitz, 1966; Helm, 1968; Muller *et al.*, 1970; Thilander and Myrberg, 1973; Maklin *et al.*, 1979; Rølling, 1980; Pilo *et al.*, 1987; Aasheim and Øgaard, 1993; Leitão, 1993; Johannsdottir *et al.*, 1997; Baccetti, 1998). It is seen more frequently in females, although gender distribution has shown some local variations when studies are compared (Helm, 1968; Magnusson, 1977; Rølling, 1980; Aasheim and Øgaard, 1993; Leitão, 1993; Johannsdottir *et al.*, 1997; Tavajohi-Kermani *et al.*, 2002).

Although there is some variation in the reported frequency of developmental dental absence (Table 1), the majority of studies indicate that the most frequently absent teeth are: the lower second premolar, followed by the upper second premolar, the upper lateral incisor and the lower central incisor (Horowitz, 1966; Thilander and Myrberg, 1973; Magnusson, 1977; Rølling, 1980; Johannsdottir *et al.*, 1997). In contrast, Muller *et al.* (1970) suggested that the most commonly absent teeth are: the upper lateral incisor, followed by the lower second premolar, the upper second premolar and the lower central incisor, and Tavajohi-Kermani *et al.* (2002) stated that the order was: the lower second premolar, the upper lateral incisor, the upper second premolar and the

lower central incisor (Table 1). Developmental absence of the first and second molars, lower canines and upper central incisors are considered relatively rare (Maklin *et al.*, 1979; Dermaut *et al.*, 1986; Schalk-van der Weide *et al.*, 1992; Guckes *et al.*, 1998).

Absence of one or two teeth is the most common finding (Muller *et al.*, 1970; Magnusson, 1977; Maklin *et al.*, 1979; Rølling, 1980; Leitão, 1993) and unilateral absence of an upper lateral incisor is often associated with malformation (e.g. microdontia) of the other lateral incisor (Magnusson, 1977; Nieminen *et al.*, 1995) (Figure 1).

Hypodontia in the primary dentition is a rare occurrence (0–0.9 per cent or < 0.9 per cent) and, when present, occurs more frequently in the incisor region, generally including the upper lateral incisor and the lower central or lateral incisor (Ravn, 1971; Bennett and Ronk, 1980; Järvinen and Lehtinen, 1981; Johannsdottir *et al.*, 1997). When hypodontia occurs in the primary dentition, (Figure 2), most authors report absence of the permanent successor in 100 per cent of cases (Ravn, 1971; Bennett and Ronk, 1980; Järvinen and Lehtinen, 1981; Johannsdottir *et al.*, 1997). However, this is not always the case (Nick-Hussein and Majid, 1996; Canut-Brussola, 2000; Dagaard-Jensen *et al.*, 1997).

Estimates of the frequency of absence of upper lateral incisors are not uniform and vary according to the population being studied (Horowitz, 1966; Muller *et al.*, 1970; Thilander and Myrberg, 1973; Magnusson, 1977; Rølling, 1980; Aasheim and Øgaard, 1993; Johannsdottir *et al.*, 1997; Tavajohi-Kermani *et al.*, 2002). Values have

Table 1 The prevalence of hypodontia in different samples and populations according to previous publications.

	Total prevalence and gender (%)	Upper lateral incisor (%)	Lower second premolar (%)	Upper second premolar (%)	Lower first incisor (%)
Aasheim and Øgaard, 1993 Norway, <i>n</i> = 1953	6.5 M 5.8, F 7.2	2			
Baccetti, 1998 Italy, <i>n</i> = 980	5.8				
Horowitz, 1966 USA, <i>n</i> = 1000	6.5	1.11	2.23	1.41	
Helm, 1968 Denmark, <i>n</i> = 1240	5 M 4.8, F 7.3				
Johannsdottir <i>et al.</i> , 1997 Iceland, <i>n</i> = 396	5 M 4, F 6	0.8	2.5		
Leitão, 1993 Portugal, <i>n</i> = 666	6.3 M < F				
Magnusson, 1977 Iceland, <i>n</i> = 1116	7.9 M 6.7, F 8.9	0.95	4.2	1.46	
Maklin <i>et al.</i> , 1979 USA, <i>n</i> = 847	4.34				
Muller <i>et al.</i> , 1970 USA, <i>n</i> = 14940	3.5	1.65	1.24	0.7	0.23
Pilo <i>et al.</i> , 1987 Israel, <i>n</i> = 702	8 M 9.7, F 6.4				
Rølling, 1980 Denmark, <i>n</i> = 3325	7.8 M 7.7, F 7.8	1.7	4.1	2.15	
Tavajohi-Kermani <i>et al.</i> , 2002 USA, <i>n</i> = 1016	8.8 M 3, F 6	1.28	1.38	0.7	0.6
Thilander and Myberg, 1973 Sweden, <i>n</i> = 5459	6.1	1.2	2.46	2	0.45

**Figure 1** Microdontia of the upper right and developmental absence of the upper left lateral incisor in a 12-year-old patient.

been reported between 0.8 and 2 per cent for the permanent dentition and between 0.1 and 0.7 per cent for the primary dentition.

Various aetiological factors have been suggested for the failure of development of the permanent tooth germ, thus leading to its absence: physical obstruction, rupture of the dental lamina, limitation of space, functional anomalies of

the tooth epithelium, initial error of the mesenchyme, or disturbances in the development of the embryonic fusion of the maxilla and the medial nasal processes (a similar situation occurs in cleft lip and palate) (Woodworth *et al.*, 1985; Svinhufvud *et al.*, 1988). It is known that the structure and development of the human dentition are rigidly controlled by genetics. Several studies in the last decades have aimed to identify those genes whose mutations may be responsible for, or associated with, hypodontia, some of which have generated important new information (Lyngstadaas *et al.*, 1996; Pirinen *et al.*, 1996; Thesleff, 1996; Vastardis *et al.*, 1996; Stimson *et al.*, 1997; Vastardis, 2000; Lidral and Reising, 2002). It is conceivable that the genes may be involved in one or more of the mechanisms proposed above for the absence of lateral incisors.

It has been hypothesized that, if hypodontia is primarily genetic, then bilateral absence would be expected to occur relatively commonly; and when hypodontia occurs unilaterally, it may be associated with structural anomalies of the contralateral tooth (such as microdontia or conical shaped teeth).

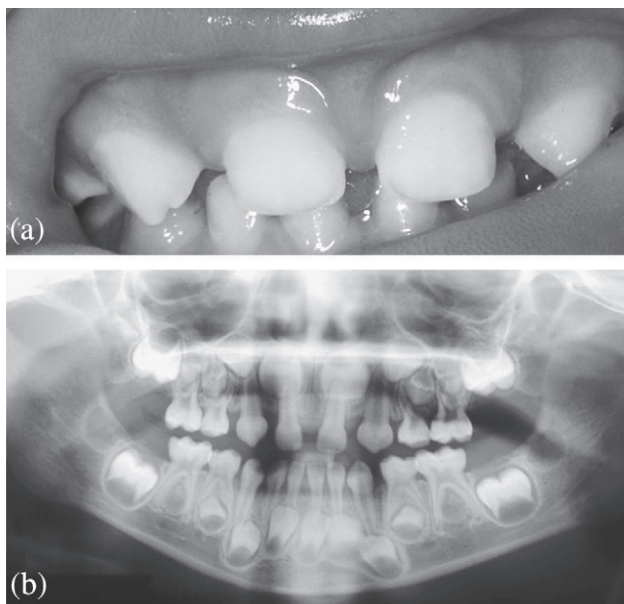


Figure 2 (a) Developmental absence of the primary upper lateral incisors in a 3-year-old child. (b) Panoramic radiograph showing the absence of the upper lateral incisors as well as of one lower incisor.

The aims of the present study were to investigate hypodontia involving missing upper lateral incisors in the Portuguese population, and to test the above hypothesis.

Material and methods

The research was performed on material from the archives of the Dentistry Clinic of the Instituto Superior de Ciências da Saúde-Norte, Portugal, using 16 771 panoramic radiographs, taken between 1993 and 2000. The patients were between 3 and 71 years of age.

The radiographs were analysed by trained observers who followed a pre-established protocol, similar to that existing in the clinic: general observation and comparison of symmetrical teeth, followed by a systematic analysis of the erupted and unerupted teeth in each quadrant, and finally of the bone adjacent to each quadrant. Radiographs showing recent extractions, bone defects, evidence of surgery, trauma and fractures were excluded.

The selection of the study sample was undertaken in two stages: first, radiographs corresponding to individuals with probable developmental absence of the upper lateral permanent incisors ($n = 267$) were selected; secondly, confirmation of developmental absence was obtained through a telephone interview or by clinical observation ($n = 219$). Demographic data including gender and age were also collected.

The study also included investigation of the following parameters: unilateral or bilateral absence, developmental absence of other teeth, supernumerary teeth, and microdontia of the contralateral maxillary lateral incisor in those

subjects presenting with unilateral absence. The terminology suggested by Proffit (1995), relating to the morphological microdontic classification of the upper lateral incisor was followed, i.e. the upper lateral incisors were considered microdontic when their mesiodistal dimension was less than that of the lower lateral incisors.

The sample was sub-divided into three groups according to age: (1) individuals younger than 8 years ($n = 12$) in whom absence of the primary upper lateral incisor was studied (data was gathered through clinical confirmation); (2) individuals who were 8 years of age and older ($n = 207$) in whom persistence of the primary upper lateral incisor related to absence of the corresponding permanent tooth was noted (data was gathered based only on radiographs, regardless of the level of root resorption of primary teeth); and (3) individuals 15 years and older ($n = 182$) in whom persistence of the primary upper lateral incisor was found.

The data was analysed using the Chi-square test. A value of $P < 0.05$ was considered significant.

Results

Of the 16 771 radiographs (8712 females and 8059 males) studied, 267 were identified as showing probable developmental absence of the maxillary upper lateral incisors. Of these, 147 (67.1 per cent) were confirmed by clinical observation and 72 (32.9 per cent) through telephone interview. Overall 219 cases were confirmed, corresponding to a prevalence of 1.3 per cent (Figure 3).

Developmental absence of the upper lateral incisor

Of the 219 patients identified as having developmental absence of the upper lateral incisor, 131 (59.8 per cent) were female and 88 (40.2 per cent) male. The prevalence was 1.5 and 1.1 per cent in females and males, respectively. This finding was significantly different ($P < 0.02$). One hundred and twenty one patients (55.2 per cent) were found to have unilateral absence and 98 (44.7 per cent) had bilateral absence ($P = 0.2$). Of those presenting with unilateral absence, 73 (60.3 per cent) were on the right and 48 (39.7 per cent) on the left side ($P = 0.06$).

Only two patients (0.26 per cent) presented with developmental absence of the primary upper lateral incisors; one had unilateral absence affecting the right side and the other had bilateral absence.

In those patients aged 8 years or over with developmental absence of the upper lateral incisors ($n = 207$), the primary tooth was retained in 14 cases (6.7 per cent). This retention was bilateral in seven individuals (3.4 per cent), and unilateral in the other seven, with the upper right being retained in four (1.9 per cent), and the upper left in three (1.4 per cent). However, in the group of patients who were aged 15 years and over ($n = 182$), only two (1.1 per cent)

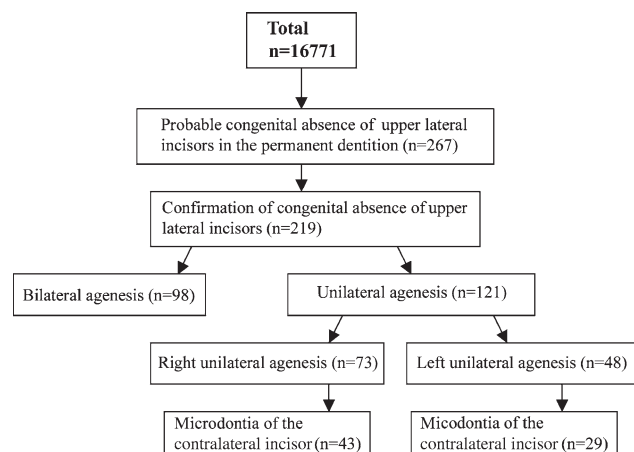


Figure 3 Analysis of 16 771 radiographs showed probable congenital absence of the upper lateral incisor in 267 subjects and of those 219 were confirmed. Ninety eight had bilateral congenital absence and 121 unilateral; 73 of these were on the right and 48 on the left side. In subjects with unilateral congenital absence, microdontia of the contralateral incisor was found in 29 subjects on the right side and in 43 on the left side.

individuals had bilateral retained primary teeth and three (1.6 per cent) retained upper left lateral primary incisors.

Other tooth anomalies associated with developmental absence of the upper lateral incisor

Microdontia of the contralateral incisor. Among the 121 patients presenting with unilateral absence, microdontia of the contralateral upper lateral incisor was found in 73 subjects (59.5 per cent), of which 29 (24 per cent) were on the right and 43 (35.5 per cent) on the left side ($P > 0.05$).

Other developmentally absent or supernumerary teeth

Of the 219 subjects identified, in only 164 was it possible to confirm the existence of other associated developmentally absent teeth. The remaining 55 individuals were excluded from this part of the analysis. Of the 164 subjects for which it was possible to confirm this information, the majority ($n = 143$, 87.2 per cent), had no other developmentally absent teeth (Table 2). For the other 21 subjects (12.8 per cent), the dental anomaly most frequently associated with absence of the upper lateral incisor was absence of the upper or lower second premolar ($n = 14$, 8.5 per cent). No supernumerary teeth were found.

Discussion

In this study, a large sample of the Portuguese population was studied in order to obtain epidemiological and clinical information related to the absence of upper lateral incisors. It is likely that data provided for this study were accurate as patients recall the extraction of teeth in this site due to the aesthetic effects. The use of panoramic radiographs allowed access to a source of reliable, extensive and easily

Table 2 Associated developmental absence of other teeth found in the present study.

Other associated congenital absences	<i>n</i>	Percentage
None	143	87.2
25	1	0.6
31	2	1.2
32	2	1.2
35	1	0.6
41	1	0.6
42	1	0.6
15, 25	3	1.8
31, 41	1	0.6
25, 35	1	0.6
35, 45	3	1.8
15, 25, 35, 45	3	1.8
15, 23, 25, 34, 35, 44	1	0.6
16, 25, 26, 35, 32, 45	1	0.6
Total	164	100.0

accessible data. However, a further clinical inquiry and/or examination was needed to confirm the absence.

Although clinical observation of all individuals would have allowed more accurate data regarding the morphology of the other incisors, as well as any other dental anomalies (Nick-Hussein and Majid, 1996; Zhu *et al.*, 1996b), this was not feasible in the current study. Therefore, as the data was mainly radiographic, the possibility of errors due to image distortion can not be excluded.

The prevalence of developmental absence of the permanent upper lateral incisors varies considerably between studies (Horowitz, 1966; Helm, 1968; Magnusson, 1977; Muller *et al.*, 1970; Rølling, 1980; Moyers, 1988; Leitão, 1993; Johannsdottir *et al.*, 1997; Baccetti, 1998; Canut-Brussola, 2000; Laskaris, 2000). In the present investigation, a prevalence of 1.3 per cent was found. This compares well with other reported values of between 0.8 and 2 per cent. Johannsdottir *et al.* (1997) quoted a prevalence of 0.8 per cent, in a group of 396 six-year old children from Iceland (average age 6 years 7 months). Horowitz (1966) found a prevalence of 1.11 per cent, in a population of 1000 individuals, with ages ranging between 7 and 16 years. Aasheim and Øgaard (1993), found one of the highest values of around 2 per cent, in a Nordic sample of 1953 nine-year-old children undergoing orthodontic examination. These differences could be related to sample selection, but it is also possible that different populations vary due to genetic variability and/or different exposure to environmental factors.

In one of the very few studies undertaken in Portugal, Leitão (1993) examined a population of 666 twelve-year-old children from the city of Lisboa and estimated a prevalence of 1.4 per cent for upper right lateral incisor absence and 1.7 per cent for upper left. In this age group, it is clearly easier to determine whether the incisor is absent or whether extraction of the incisor has occurred. In the present study, in order to overcome this limitation, individual patients

were contacted by telephone, so that those subjects where the tooth had been extracted could be excluded. The present findings cannot be compared with the results obtained by Leitão (1993), since the prevalence of upper lateral incisor agenesis was determined only for a subset of the population and, in addition, unilateral versus bilateral absence was not investigated, nor the association of upper lateral incisor absence with other dental anomalies. However, for the data that can be compared, the prevalence values found were within the same age range, and the gender distribution also appears similar. The only difference is that the upper left lateral incisor was noted to be absent more often in Leitão's study. The sample obtained in Lisboa is likely to be more variable in terms of ethnicity than in the present sample and this could explain some of the differences.

Muller *et al.* (1970) reported a prevalence of 1.65 per cent for agenesis of the upper lateral incisor, this value being similar to that in the present study, in a sample of 14 940 North Americans (13 459 Caucasians and 1481 Afro-Americans), aged between 6 and 14 years. Those authors also found, interestingly, that when one or two teeth were absent, the upper lateral incisor was the most frequently absent, but when more than two teeth were absent, the second premolar was the most frequently absent.

Developmental absence of the upper lateral incisors has been shown to be more common in females (Horowitz, 1966; Helm, 1968; Magnusson, 1977; Muller *et al.*, 1979; Rølling, 1980; Dermaut *et al.*, 1986; Leitão, 1993; Johannsdottir *et al.*, 1997; Laskaris, 2000), which is in agreement with the results of the present study, where an important difference in gender distribution ($P < 0.02$) was found. The reasons for this consistent gender difference are not clear, and may be related to gender-determined features, i.e. sexual dimorphism in general skeletal growth and in tooth eruption (Proffit, 1995).

Of the patients under 8 years of age, only two (0.26 per cent) had developmental absence of the primary lateral incisors, one being bilateral. This confirms that the occurrence of hypodontia in the primary dentition is rare (Ravn, 1971; Bennett and Ronk, 1980; Järvinen and Lehtinen, 1981; Johannsdottir *et al.*, 1997) and, when verified, the permanent successors were also absent (Ravn, 1971; Järvinen and Lehtinen, 1981). It must be noted, however, that other studies (Nick-Hussein and Majid, 1996; Canut-Brussola, 2000; Daugaard-Jensen *et al.*, 1997) have shown that absence of a primary incisor is not always followed by that of the succedaneous tooth.

In the patients aged 8 years or over, the primary tooth was retained in 6.7 per cent. However, among patients aged 15 years or over, only 2.7 per cent presented with retention of the primary tooth. Therefore, retention of the primary lateral incisors as an aesthetic replacement in the absence of the permanent lateral incisors is not feasible in many cases.

The results of this study are in agreement with those of other authors (Magnusson, 1977; Rølling, 1980; Aasheim and Øgaard, 1993), who also found unilateral absence was more common than bilateral absence, with a trend for the upper right lateral incisor to be absent more often than the left, although the difference was not statistically significant. In subjects with clefts of the lip/palate, an asymmetrical distribution has been described, but with the left side more often affected (Tsai *et al.*, 1998; Shapira *et al.*, 2000); this has weakened the hypothesis that developmental tooth absence and cleft lip/palate could result from similar mechanisms (Ranta *et al.*, 1983; Ranta and Tulensalo, 1988). In addition, studies of the relatives of children with cleft lip/palate did not show an increased frequency of developmental absence (Anderson and Moss, 1996).

In the present study all teeth where the mesiodistal dimensions were reduced were classified as microdont. In agreement with Horowitz (1966), Svinhufvud *et al.* (1988), Baccetti (1998) and Nieminen *et al.* (1995), microdontia was the anomaly most frequently associated with hypodontia. Of the 121 patients in this study who had unilateral absence, 72 (59.5 per cent) were found to have a microdont lateral incisor on the other side. Nieminen *et al.* (1995) proposed that microdontia of one or more upper lateral incisors represented variable expression of the same anomaly. The data in the present study supports their theory, as does the investigation by Garn and Lewis (1970), who stated that microdontia is associated with missing teeth, and is more evident in cases of multiple agenesis and occurs more frequently in females than in males. Schalk-van Der Weide *et al.* (1994), stated that in patients presenting with developmentally missing teeth, the mesiodistal dimensions and the labiolingual dimensions are diminished for all teeth. In a later study, Schalk-van der Weide and Bosman (1996) also noted a reduction in the size of some teeth in relatives of patients presenting with hypodontia.

The terms 'hypodontia' and 'concomitant hyperdontia' (Segura and Jimenez-Rubio, 1998) and 'oligo-pleiodontia' (Nathanil, 1970) have been used to describe a situation in which developmental absence and supernumerary teeth are present in the same individual. These situations are rare (Ranta and Tulensalo, 1988; Zhu *et al.*, 1996 a,b), and were not found in the present study. This confirms the results of Baccetti (1998), who stated that supernumerary teeth appear to be of a separate aetiological entity from the absence of upper lateral incisors. As the present sample was larger than those quoted, it may be concluded that this situation occurs even less frequently in the Portuguese population. However, the possibility of these results being distorted by possible extraction of the supernumerary teeth cannot be completely excluded.

The aetiology of hypodontia remains unknown in spite of recent progress. It is interesting that the teeth that erupt in critical terminal areas of the dental lamina (such as the upper lateral incisor, second premolars, third molars) and those

located in the embryonic fusion areas, (the latter growing within the tissue corresponding to each 'family' of teeth) are the teeth that are most frequently affected, the so-called 'end of series' anomaly (Svinhufvud *et al.*, 1988; Thesleff, 1996). Sejrsen *et al.* (1995) and Kjær (1997) explained agenesis as a product of the lack of innervation in the final stages of development on the three distinct bilateral neural fields, thus altering the development of the teeth furthest from the innervation of the field. In agreement with this, there is a close association between developmental absence of the second premolars and absence of the upper lateral incisors. Molecular factors that control neural development may also have an impact on tooth formation and therefore their malformation may lead to failure of development. The continued search for the genetic basis of hypodontia may help to elucidate these mechanisms.

Conclusion

The findings showed that 1.3 per cent of the individuals studied had developmental absence of the upper lateral incisors, with females being more frequently affected. The most common presentation was that of unilateral absence of the upper right lateral incisor associated with microdontia of the contralateral incisor, suggesting the possibility of the existence of a variant expression of the same characteristic.

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